

# Verrucous Carcinoma of the Lower Lip: A Case Report

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Verrucous carcinoma is a relatively rare variant of well differentiated squamous cell carcinoma first described by Ackerman in 1948. It is distinct in its slow progression, exophytic cauliflower like growth, low grade malignancy and low incidence of metastasis. The oral cavity is one of the predilection sites for verrucous carcinoma. In the oral cavity, the gingiva and buccal mucosa are the common site. Verrucous carcinoma of the lip is clinically quite rare and only several cases of that were reported in the world. The aim of this study is to report an unusual case of verrucous carcinoma of the lower lip.

**Key Words:** Lip; Verrucous carcinoma

## INTRODUCTION

Verrucous carcinoma is an unusual, non-metastasizing, distinct variant of well differentiated squamous cell carcinoma. The term verrucous carcinoma was first described by Ackermann<sup>1)</sup> in 1948, hence the tumor is also known as “verrucous carcinoma of Ackermann” or “Ackermann’s tumor”.<sup>2)</sup> The oral cavity is the most common site of occurrence for verrucous carcinoma.<sup>2)</sup> However, oral verrucous carcinoma is a rare tumor, diagnosed in only 1 to 3 of every 1,000,000 persons each year.<sup>3,4)</sup> In addition, verrucous carcinoma of lips is considered as extremely rare and few studies are available. We describe our experience with this unusual case of verrucous carcinoma of the lower lip.

## CASE REPORT

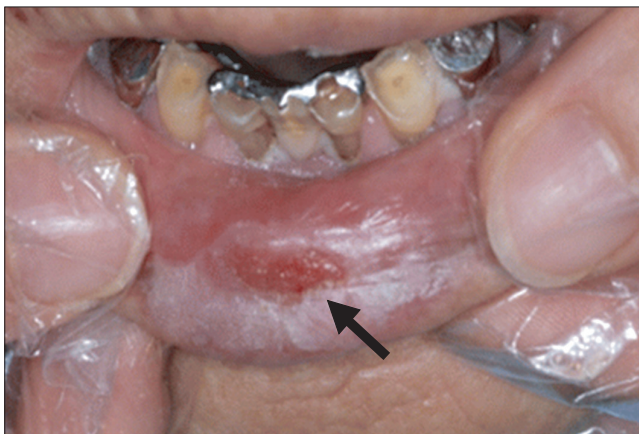
In September 2012, a 75-year-old female patient presented to the Department of Oral Medicine at the Chonbuk National University Hospital (Jeonju, Korea) with a complaint of the swelling and pain of the lower lip for about three years. The lesion of lower lip had been treated at a

dermatologic clinic, but the regimen was not effective. She has no relevant history except for hypertension.

On clinical examination, there was a whitish hyperkeratotic lesion on the right lower lip with pain (Fig. 1). No lymphadenopathy was detected on the neck or sublingual area. The punch biopsy was performed in company with laser treatment and the biopsy specimen was reported as mild to moderate epithelial dysplasia. After three weeks of laser



**Fig. 1.** Whitish hyperkeratotic lesion on the lower lip at the first visit.



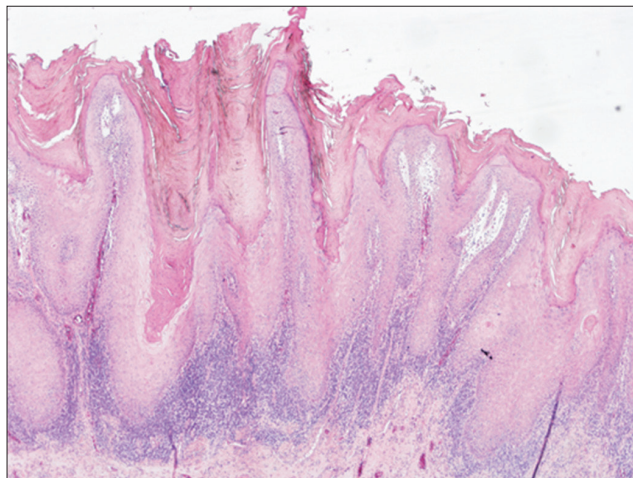
**Fig. 2.** Unhealed and erosive wound after 3 weeks of laser treatment showing bleeding tendency (arrow).



**Fig. 4.** Erosive lesion in the center of hyperkeratotic lesion of the lower lip after 3 months of treatment (arrow).



**Fig. 3.** Recurrent hyperkeratotic lesion without erosion after 6 weeks of treatment.



**Fig. 5.** Microscopic findings of operation specimen showing marked keratosis and downward invasion of epidermal keratinocytes with a blunt border and a pushed-down appearance (H&E staining, x100).

treatment, the treated lesion was not healed and showed erosion with bleeding tendency (Fig. 2).

In December 2012 (after 6 weeks of treatment), the erosion was disappeared, but the hyperkeratotic lesion was recurrent. And then mild paresthesia on the area of laser treatment was presented (Fig. 3).

In January 2013 (after 3 months of treatment), new erosive lesion was developed in the center of hyperkeratotic lesion (Fig. 4). She was referred to Department of Oral and Maxillofacial Surgery for more evaluation and removal of the lesion.

In March 2013, incisional biopsies were performed at the boundary of the lesion and the biopsy specimens were reported as mild to moderate epithelial dysplasia.

In April 2013, surgical excision of the lesion was performed under general anesthesia for definitive diagnosis

and removal of the lesion. Microscopic examination of the surgical specimen revealed the presence of marked proliferation with down growth of epithelium into the connective tissue without invasion (Fig. 5) and was confirmed verrucous carcinoma.

## DISCUSSION

Verrucous carcinoma is a rare form of well differentiated squamous cell carcinoma. This tumor is distinct in its slow growth and no metastatic potential.<sup>2,4)</sup> But this lesion can be locally destructive despite its deceptively benign clinical behavior if not treated.<sup>1,5)</sup> It is predominantly seen in elderly males over the sixth decade.<sup>6,7)</sup> Ackerman<sup>1)</sup> first described

this tumor of the oral mucous membrane, which is now also known as “verrucous carcinoma of Ackermann” or “Ackerman’s tumor”.<sup>2)</sup> The term verrucous is used because of its exophytic squamous mucosal or cutaneous tumors that are heaped above the epithelial surface with a papillary micronodular appearance.<sup>5)</sup>

This tumor may occur in several locations in the head and neck with the most common sites being the oral cavity (55.9%).<sup>5,8)</sup> Extra-oral sites of occurrence are larynx, esophagus, paranasal sinus, skin, penis, vulva, uterine cervix and it also from odontogenic cyst lining.<sup>4)</sup> The most common site of the tumor in the oral cavity is gingiva followed by buccal mucosa.<sup>5)</sup> Verrucous carcinoma of the lip is very rare. In 1981, lip verrucous carcinoma was first introduced by Diaz-Pérez et al.<sup>9)</sup> Because it is unusual and can be difficult to diagnose, the treatment may be delayed.

Verrucous carcinoma is usually exophytic papillary in nature with pebbled surface. But it may always not appear as typical appearance. In this case, the patient presented with a hyperkeratotic lesion without papillomatous surface.

The etiology of verrucous carcinoma is not completely established, though a few studies have shown an association with earlier injuries and scars, as well as with chronic inflammation.<sup>10,11)</sup> Tobacco including snuff use is also associated with development of verrucous carcinoma.<sup>2-4)</sup> Verrucous carcinoma is so closely aligned with the use of snuff and chewing tobacco that it has been called the “snuff dipper’s cancer”.<sup>3)</sup> There was no history of trauma or tobacco in our case. Human papillomavirus (HPV) has also been implicated as the cause for verrucous carcinoma.<sup>12-15)</sup> Although the role of human papilloma virus in the etiology of verrucous carcinoma is still unclear, HPV-deoxyribonucleic acid (DNA) types 6, 11, 16, and 18 has been identified by polymerase chain reaction (PCR), restriction fragment analysis, and DNA slot-blot hybridization in verrucous carcinomas.<sup>12-14)</sup> However, HPV has not consistently been identified in all such carcinomas.<sup>16)</sup> It is regrettable for us not to have checked HPVs in our case. Further studies must be required to clarify the etiology of verrucous carcinoma.

Microscopically this tumor is characterized by predominant exophytic overgrowth of well-differentiated keratinizing epithelium having minimal atypia associated with intense chronic inflammatory infiltrate.<sup>1,7)</sup> The pathological

diagnosis of verrucous carcinoma is not so difficult. But in some cases, small or superficial biopsy specimens will show only hyperkeratosis, acanthosis and papillomatosis.<sup>4,17)</sup> In our case, first and second biopsy specimens were just diagnosed as epithelial dysplasia. Because verrucous carcinoma is a benign-appearing lesion with “pushing” borders, and the basement membrane can appear to be intact, it can be mistaken as a benign lesion histologically.<sup>8)</sup> Deeper adequate biopsy and sufficient volume of biopsy specimen must be taken whenever the clinician suspects verrucous carcinoma for definitive diagnosis. However, in oral region, it is often difficult technically to obtain enough amount of the specimen. In fact, multiple biopsies may be required to confirm the diagnosis of verrucous carcinoma.<sup>18,19)</sup>

Once a diagnosis of verrucous carcinoma has been established, complete resection of the tumor is considered as the treatment of choice.<sup>10,20)</sup> In almost all cases of verrucous carcinoma, neck dissection is not necessary because lymph node metastases are extremely rare.<sup>17)</sup> Verrucous carcinoma has a good prognosis when treated early with local excision; Extensive lesions, however, may require more-aggressive procedures.<sup>19)</sup> Finally, it is important to diagnose accurately the verrucous carcinoma for preventing the most extensive involvement of adjacent areas and/or wide surgical resection of the tumor.

## CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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